

SPEECH AND PROSODY IN
DEVELOPMENTAL DISORDERS: AUTISM
AND DOWN'S SYNDROME

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Abstract

Language impairment is a key characteristic of many developmental disorders, with the relationship between linguistic and cognitive ability a critical topic for research in this field. Speech (articulation and phonology) and prosody have largely been absent from these discussions, perhaps because they are not universally impaired. The portfolio of published research critically appraised here addresses the relationships between speech and prosody and other domains, such as language and cognition, in two conditions in which disordered speech is common: primarily at the suprasegmental level in autism and at the segmental level in Down's syndrome.

Speech disorders were found in both conditions, though speech was much more severely impaired in Down's syndrome. Errors were typically categorised as delayed phonological processes, implying a linguistic cause. However, through fine phonetic transcription and instrumental techniques it was shown that both conditions also presented with distortions that were more phonetic in nature and with non-developmental errors. Severity of speech disorder was not related to cognitive or linguistic ability as measured by standardised assessments, suggesting that a generalised delay in language or cognition was not the cause of disordered speech. In autism minor delays and distortions may be due to a lack of ability to identify with peers and impaired theory of mind, whereas in Down's syndrome anatomical differences and difficulty with motor planning are likely causes. Both linguistic and paralinguistic prosody were found to be disordered in children with autism and correlations with linguistic ability were found. However, disordered prosody is more likely to be due to impaired theory of mind or weak central coherence than a result of delayed language.

Both autism and Down's syndrome present with speech that is disordered rather than simply delayed and this is unlikely to be due to delayed language, suggesting that specific, targeted intervention may be warranted.

Keywords: Prosody, speech, autism, Down's syndrome.

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Preface

The critical appraisal that follows, together with a selection of my publications, is the culmination of around seven years of research. Over this time my research has addressed the relationships between speech and other domains, such as language and cognition, in developmental disabilities. I have selected six publications as representative of this theme (referred to as papers 1 to 6), but these papers are not an exhaustive list of my output on this topic. Other papers are therefore referred to in the traditional manner and referenced in the reference list. Papers prior to 2009 are under my previous name of McCann.

Paper 1 (McCann & Peppé, 2003): A critical review, provides an overview of the literature on prosody in autism. Research published since 2003 is evaluated in the critical appraisal.

Papers 2 (McCann et al., 2007) & 3 (Peppé et al., 2007): Two papers on the main results of my research on prosody in autism. Paper 2 covers mainly the relationship between language, cognition and prosody and paper 3 compares the prosodic skills of children with autism to typically developing children and adults.

Paper 4 (McCann et al., 2008): An illustration of the heterogeneity of prosodic disorders in autism through two case studies.

Paper 5 (Cleland et al., 2010): Key results of my research on types of speech disorders in Down's syndrome and how they relate to linguistic and cognitive skills.

Paper 6 (Cleland et al., 2009): Electropalatographic data from the speech of children with Down's syndrome, highlighting the need to take fine phonetic differences into account when diagnosing speech disorders.

Selected Publications

1. McCann, J. & Peppé, S. (2003). Prosody in Autism Spectrum Disorders: A Critical Review. *International Journal of Language and Communication Disorders*, 38, 325-350.
<http://informahealthcare.com/doi/pdf/10.1080/1368282031000154204>
2. McCann, J., Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Prosody and its relationship to language in school-aged children with high-functioning autism. *International Journal of Language and Communication Disorders*, 42, 682-702.
<http://dx.doi.org/10.1080/13682820601170102>
3. Peppé, S., McCann, J., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Receptive and expressive prosodic ability in children with high-functioning autism. *Journal of Speech, Language, and Hearing Research*, 50, 1015-1028.
<http://jslhr.highwire.org/cgi/reprint/50/4/1015>
4. McCann, J., Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2008). The Prosody-Language Relationship in Children with High-Functioning Autism. In McGregor, E., Nunez, M., Cebula, K. and Gomez, J.C. (Eds.) *Autism. An Integrated View from Neurocognitive, Clinical, and Intervention Research*. Oxford, United Kingdom: Blackwell, 214-235.
<http://ereseach.qmu.ac.uk/1544/>
5. Cleland, J., Wood, S., Hardcastle, W., Wishart, J. & Timmins, C. (2010). The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. *International Journal of Language and Communication Disorders*, 45, 83-95.
<http://dx.doi.org/10.3109/13682820902745453>
6. Cleland, J., Timmins, C., Wood, S., Hardcastle, W. & Wishart, J. (2009). Electropalatographic therapy for children and young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23, 926-939.
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1 Introduction

Developmental disorders are common and varied. Many of these disorders are diagnosed based on the results of genetic testing, for example Down's syndrome (DS). In other cases, such as specific language impairment (SLI) or autism spectrum disorders (ASD), diagnosis relies on clinical observation and exclusion criteria. While the aetiologies of these disorders may be different, they all share impairments in language. The study of linguistic ability (and its relation to cognitive ability) in developmental disorders has long been a critical topic for research in this field. However, speech (articulation and phonology) and prosody have largely been absent from these discussions. This is perhaps because although disordered speech or prosody are key features in some developmental disorders they are not universally impaired.

Two of the most frequently studied developmental disorders are DS and ASD. Both feature disordered speech, primarily at the segmental level in DS (papers 5 and 6, Cleland, Wood et al., 2010 and Cleland et al., 2009) and the suprasegmental level in ASD (papers 1, 2, 3 and 4, McCann & Peppé, 2003; McCann et al., 2007; Peppé et al., 2007 and McCann et al., 2008), but prior to our research it was not clear whether these speech disorders are related to a more general cognitive or language delay or whether non-linguistic causes are more likely. Intelligibility is a major issue in DS (Rondal & Edwards, 1997) yet it is rarely studied in the context of the cognitive and language impairment that is universal in DS. Similarly, disordered prosody is a key aspect of ASD, featuring in Kanner's original description of autism (1943). It may differentiate ASD from other developmental disorders, yet it is hardly studied at all, perhaps because it is difficult to measure or because impairments in prosody may be paralinguistic in nature (Peppé, 2009). Again, it is not clear whether disordered prosody is related to language impairment in autism, or whether other explanations, such as differences in cognitive style, are more likely. In our research we show how speech and prosody problems in developmental disorders can be due to factors outwith narrow linguistic constraints, offering anatomical and physiological explanations in DS and social and cognitive explanations in ASD.

1.1 Speech and Prosody in the Context of Linguistic Impairment

The study of developmental disorders aims to link behavioural symptoms to biological causes (Morton, 1994). DS has a clear biological cause (usually trisomy 21) and it is generally accepted that ASD has biological causes, even if these have not yet been

identified. However, prior to our papers very little research had attempted to link disordered prosody to either the biological or cognitive level in ASD, with the exception of one study by Rutherford et al. (2002) on affect and theory of mind (see 2.2.2 and 2.2.7.1). Research on speech in DS has focused on impairments in the phonological system (at the cognitive level) but speech development in DS is complicated by anatomical and physiological differences (Spender et al., 1995; Miller, Leddy & Leavitt, 1999). However, most studies investigating speech production in DS take a narrowly linguistic view, using broad transcription to report delayed phonological processes (e.g. Stoel-Gammon, 1980; Van Borsel, 1996). This is problematic since fine phonetic differences, perhaps with anatomical and physiological causes, are not recorded, implying that the speech of people with DS sounds much like that of younger typical children when in fact it is much less intelligible and is phonetically different. Describing impaired speech in terms of phonological processes implies a deficit in the phonological system when this may not be the case. For example, limited phonotactic forms may be due to structural processes, such as final consonant deletion, or may be explained by impairments in motor programming. At the very least, where speech disorders are complex, as they are in DS, then narrow transcription should be used (paper 5); although fine phonetic differences are even more likely to be apparent with instrumental techniques (paper 6).

Prosody has an interesting function in communication, having both linguistic and paralinguistic functions (Crystal, 1969). Prosody signals syntactic boundaries and word stress, which are clearly linguistic, and therefore we might expect a relationship between these types of prosody and other linguistic skills such as syntactic ability. On the other hand, the conveying of emotion and pragmatic uses of prosody such as contrastive stress are paralinguistic and therefore might be less likely to be related to core linguistic skills. Logically, types of prosody that are clearly linguistic might be impaired in developmental language disorders. In contrast, since social interaction, pragmatics and affect are known to be impaired in ASD we might expect paralinguistic functions of prosody to be specifically impaired in ASD. Our research (papers 1, 2, 3 and 4) therefore looks at each type of prosody in turn, looking for cognitive explanations for impairment.

1.2 Using Standardised Tests to Investigate Speech and Prosody

Most large studies of speech, language and cognitive skills in developmental disorders use standardised tests. This has several advantages. Performance is compared to a large number of typically developing peers without the need for a control group. Clinicians are usually familiar with these tests and have access to them, allowing them to use the same tests

with their own clients. However, problems arise with the use of standardised assessments of speech in developmental disorders. Speech development (as measured by accuracy of consonants in single words) is not normally distributed in the school-aged population, since speech development is largely complete by early primary school (Shriberg, Tomblin & McSweeney, 1999), as well as being highly variable. Some tests (for example, the Goldman-Fristoe Test of Articulation-2, Goldman & Fristoe, 2000) are standardised across a wide age range (2 to 21 years), but scores are not normally distributed and therefore not truly comparable with standard scores from language and cognitive assessments. Other tests, such as the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd et al., 2002) are standardised only up to age seven when most children achieve ceiling scores on single word phonology/articulation tests. Most studies tend to report standard scores for these tests in the same way as they report standard scores for language or cognitive tests. In studies of ASD this has led to the conclusion that articulation skills are spared (Kjelgaard & Tager-Flusberg, 2001). Comparing speech development with cognitive or language development with standardised assessments is therefore more complex than it might appear.

Studying prosody is even more complex since there are no standardised tests (Diehl & Paul, 2009). This lack of assessment tools may be because prosodic disorders are less common than speech or language disorders or it may be because prosodic disorders are thought of as hard to define and relatively little time is devoted to their study in the training of speech and language therapists (Peppé, 2009). In our research we used a non-standardised test, the PEPS-C (Peppé & McCann, 2003) and collect carefully matched control data.

2 Autism Spectrum Disorders

Autism is a triad of impairment: atypical social interaction; atypical communication; and restricted, stereotyped and repetitive behaviours (Wing & Gould, 1979). Although disordered communication is only one third of the triad, deficits in this area are the most frequently observed characteristic of autism (Whitehouse et al., 2008) and preschool language ability predicts later cognitive, linguistic and adaptive functioning (Venter et al., 1992). Moreover, autistic symptomology correlates with linguistic impairment, that is, formal aspects of language such as performance in tests of pure grammar (Whitehouse et al., 2008). Language is always delayed and may be disordered in preschool children with autism but formal aspects of language may be in line with peers in older children and adults. Most research has focused on pragmatic and social aspects of language, showing difficulties in these higher-order aspects of language processing (Tager-Flusberg, 1996). Core linguistic

skills, however, are much more variable, ranging from complete absence of expressive language to fluent speech with large vocabularies.

2.1 *Speech in Autism*

In contrast to delayed and disordered language, articulation and phonology are reported to be either age-appropriate or superior to other expressive language abilities (Rapin & Dunn, 2003). In Kjelgaard and Tager-Flusberg's (2001) study of 89 children with autism, articulation was described as "spared". Rapin et al. (2009) used standard scores from an articulation test to drive cluster analysis of language abilities in 62 children with autism. They proposed two types of language disorders: severe impairment in expressive phonology (24%) and borderline/normal phonology with impaired comprehension (76%). The suggestion that nearly a quarter of children with autism present with impaired phonology is striking and at odds with the Kjelgaard and Tager-Flusberg (2001) study.

A closer look at methodology suggests that both of these studies have questionable results. First, they used single word articulation tests which were scored only on a right/wrong basis. Second, they mixed phonetic and phonological impairments, for example, a lateral /s/ would be scored as incorrect although it is not thought of as a phonological impairment. Lastly, in tests such as these we would expect ceiling scores in the school-aged population, at least over the age of seven, but both studies report that many children made a small number of errors. In children of this age even a small number of errors can constitute a significant speech disorder.

Like the Kjelgaard and Tager-flusberg study, most (84%) of the children in our study (paper 2) presented with standard scores in the normal range in an articulation test. However, a critical re-examination of our original paper prompted by the issues raised here reveals that only 14 (45%) children made no errors at all on the GFTA, despite being over seven years. This is an unexpected finding, suggesting that subtle speech impairments may be evident in many children with autism and highlighting the need to look beyond standard scores. Further analysis of a larger group (the same children with HFA plus a further 39 children with Asperger's syndrome, now published in Cleland, Gibbon et al., 2010) showed that the most common processes in children with ASD were developmental (shown in Figure 1 with developmental errors in white and non-developmental errors in black).

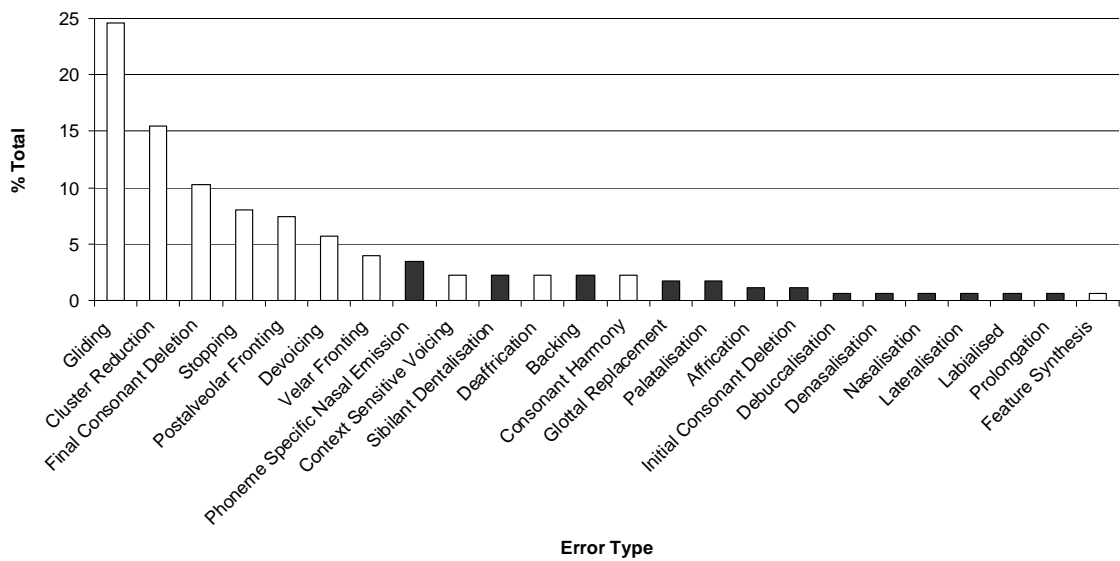


Figure 1: Relative occurrence of a range of speech error types in children with ASD (Developmental errors are shown in white and non-developmental errors in black).

Some errors appeared in individual children only, and of these three were developmental (stopping, velar fronting and context sensitive voicing). Interestingly, three children produced non-developmental errors. Backing is a phonological error resulting in loss of contrast between alveolar and velar stops, affecting intelligibility. Phoneme specific nasal emission and dentalisation of sibilants are phonetic distortions, likely to affect the social acceptability of speech. The children who produced these latter acceptability errors made no other types of errors. These findings echo those of Shriberg et al. (2001) who report what they call “residual articulation errors”: dentalised sibilants, derhoticisation (for American speakers), lateralised sibilants and labialised /l/, in 30 adolescents and adults with ASD. These types of errors, along with minor developmental errors such as gliding, are unlikely to affect intelligibility significantly but may diminish the social acceptability of speech. Evidence for this is found in studies of (otherwise typically developing) children with minor articulation disorders. Hall (1991) found that primary school children showed negative attitudes towards their peers with mild articulation disorders (/r/ or sibilant distortions). In people with ASD who already have difficulty with social interaction, the presence of even a mild articulation disorder is likely to compound the problem.

Shriberg et al. (2001) do not suggest why minor articulation errors are likely to persist in ASD. In our study of children (Gibbon et al., 2004) we suggest that speech impairments, especially delayed phonological processes, may be related to delayed language.

Though paper 2 did not find any correlation between language skills and articulation that is not to say that early language delay (present in autism but not Asperger syndrome) does not lead to delayed phonology. Why delayed phonology persists and why phonetic distortions are found in adults with ASD in the face of normal formal language skills is not so easily explained by a general delay in language. In Baron-Cohen and Staunton's study of accent acquisition (1991), children with non-native mothers were more likely to develop her foreign accent than the ambient native accent. They suggest that children with autism lack the drive that typical children have to identify with peers, and so do not develop the appropriate accent. In the case of speech disorders, people with autism do not (presumably) receive a disordered model from their parents. However, it is possible that when distortions or minor delays exist, people with autism have less drive to change the errors since they identify less with their peers. This might especially be the case with minor distortions or delays such as gliding, where errors do not affect intelligibility. Since the person with autism is successful at conveying their message, any desire to change errors would have to be motivated by a drive to produce more socially acceptable speech. This is unlikely to occur in ASD, perhaps because people with autism may lack the theory of mind (see 2.2.7.1) to even appreciate that other people perceive their speech as unusual or different from the norm.

2.2 *Prosody in High-Functioning Autism*

Paper 1 shows that very little research exists quantifying the expressive prosodic disorder in autism and even less research investigates receptive prosody. Papers 2, 3 and 4 therefore set out to investigate prosodic disorder in ASD with a view to describing the disorder and relating it to cognitive and language skills. There are several different ways of categorising expressive prosodic disorders (Peppé, 2009) but for our purposes they broadly fall into two categories. Disorders of function result in speech that is unable to convey important distinctions using prosody alone. For example, a speaker may aim to produce a sentence such as "John plays football" either as a question or statement (illocutionary force, a pragmatic use of prosody) but if they do this with the same type of prosody, the intended functions will be indistinguishable. In contrast, disorders of form, or overt prosodic disorders, result in speech that is unusual sounding, such as exaggerated intonation, but still able to convey functions. Of course, many disorders of form may also result in the speaker being unable to use prosody functionally. Additionally, receptive prosodic disorders may lead to difficulties understanding another person's intentions through prosody, for example distinguishing a question from a statement or identifying emotions.

2.2.1 *Measuring Prosody in Autism*

In our studies (papers 1, 2, 3 and 4) we took a comprehensive look at prosody using the psycholinguistic PEPS-C assessment (Profiling Elements of Prosodic Systems in Children, McCann & Peppé, 2003, a full description of the test is available in the appendix of paper 3). One weakness of PEPS-C is that it does not cover all aspects of Stackhouse and Wells (1997) psycholinguistic framework. Appendix One therefore shows which levels are tested in the PEPS-C assessment, and additionally proposes new tasks for levels not tested.

2.2.2 *Affective Prosody*

Affective prosody has been described as “non-linguistic” (Pell & Baum, 1997) and we might therefore expect performance in these subtests to be independent of core linguistic skills. However, prosody can only be realised in combination with segmental, lexical and syntactic information (Seddoh, 2002) making affective (and pragmatic) prosody better described as paralinguistic. Seddoh (2002), suggests that the presumed dichotomy between affective and linguistic prosody is misleading, and that the two are intertwined, suggesting that it might be possible to find some relationship between paralinguistic prosody and core linguistic skills. Deficits in paralinguistic aspects of prosody may indicate a more specific prosodic deficit, rather than generalised language impairment. Since affect and pragmatics are known to be impaired in autism we might expect deficits in these aspects of prosody.

In our studies (papers 2, 3 and 4) children with autism showed deficits in both the understanding and use of affective prosody. Since PEPS-C tests only a basic affective distinction (liking versus disliking), and moreover a distinction which is early acquired by typical children (73% of 5 year olds pass the affect reception task, rising to 89% at 6 years and 100% at 7 years) this suggests a fairly severe difficulty with affective prosody. However, this conclusion is problematic since PEPS-C tests only two emotions. Most research in emotion recognition (mainly from facial expression) focuses on six basic emotions: happiness, sadness, fear, anger, surprise and disgust (Ekman, 1999), PEPS-C therefore essentially tests only the distinction between happiness (liking a food) and either sadness (not wanting to eat a certain food) or disgust (disgusted by the idea of eating a certain food). Future research into affective prosody should test at least all of the basic emotions, progressing to investigating more complex emotions. Clearly the expression and reception of prosodic affect in autism is an important area for future research, especially since people with autism show difficulties with emotion in other modalities, such as facial expression (Celani, Battacchi & Arcidiacono, 1999). Paul et al. (2005) did not find a deficit in affect in

their study of older individuals with ASD, but again they test only limited distinctions, in this case “anxious” (fear) versus neutral. Paul et al.’s results are difficult to interpret due to ceiling effects, but perhaps suggest that there continues to be development in both the understanding and use of affective prosody across the lifespan in ASD.

In paper 2 we presented only correlations between language and receptive, expressive, function and form of prosody, not individual subtests. To further explore the themes developed here, the data is reanalysed to determine whether paralinguistic as well as linguistic aspects of prosody correlate with core linguistic skills. Table 1 shows these new results. Since multiple tests were required a Bonferroni correction was applied (Curtin & Shulz, 1998). Despite prosodic affect being paralinguistic, understanding of this function of prosody correlated highly with receptive vocabulary and expressive language. This is not explained by a correlation with chronological age, or non-verbal ability. From this it is tempting to conclude that poor language skills cause difficulty understanding affect, however, the correlation between the two measures is not necessarily an indication of causality (Coolican, 2009). It is also a possibility that a difficulty with reciprocal relationships and therefore affect leads to a difficulty learning language. Or, the correlation may be explained by a third factor. Rutherford et al. (2002) suggest that understanding of affect is essentially a Theory of Mind (ToM) task (see 2.2.7.1). Since ToM is known to correlate highly with language skills (Astington & Jenkins, 1999) it is possible that the correlation between the affect tasks and linguistic skills may be due to a further correlation with ToM. Further evidence for this comes from a dissociation between language and performance in the expressive affect task, but again the usefulness of this result is limited since only one type of affective prosody was investigated. Section [2.2.6.1](#) explores the relationships between prosody, language and ToM further.

Table 1 : Correlations between PEPS-C and language and cognitive measures.

	BPVS (receptive vocabulary)	TROG (receptive grammar)	CELF (expressive language)	GFTA (articulation)	RM (non-verbal ability)	Age
PEPS-C Task						
Turn-end Input	r=.630; p<.0007					
Turn-end Output			r=.678; p<.0007			
Affect Input	r= .597; p<.0007		r=.684; p<.0007			
Affect Output						
Chunking Input		r=.630; p<.0007				
Chunking Output						
Focus Input						
Focus Output		r=.718; p<.0007				
Intonation Input						
Intonation Output						
Prosody Input						
Prosody Output						

Note: Only correlations significant at the p<0.0007 level (Bonferroni corrected) are reported. Blank cells are not significant

2.2.3 *Pragmatic Prosody*

Peppé (2009) describes stress as straddling affective and grammatical functions and as such we might expect it to be impaired in ASD, and especially so in autism where language development is delayed. Early studies of prosody in autism focused on contrastive and default stress (see paper 1), finding both to be problematic (Baltaxe, 1984 and Baltaxe & Guthrie, 1987). Yet, in paper 3 we found no differences in the understanding of contrastive stress between children with and without autism, with floor effects in both groups. However, in the expression task children with autism were more likely than the control group to place stress early in the utterance. Previous studies of contrastive stress in typical children have shown that, like our control group, expression of contrastive stress is acquired relatively early. Hornby and Hass (1970) found that children as young as 3;0 to 5;11 were able to use contrastive stress in corrections. In contrast, comprehension of contrastive stress is acquired relatively late, with Wells, Peppé and Goulondris (2004) finding that only 13 year olds were competent at a task similar to ours. Since the children in our studies were younger than this, it is not surprising that they found the understanding of contrastive stress so problematic. It would therefore be useful to look at contrastive stress in an older group of children with autism in order to determine whether the children are presenting with a pattern of delay.

In our other task of pragmatic prosody, turn-end, we again found no differences between groups due to floor effects. However, in the expression task children with autism produced more statement-type responses, suggesting that they were unable to signal questions using prosody alone. Again, a correlation with receptive vocabulary was evident. However, the PEPS-C turn-end task is problematic in that in spontaneous conversations questioning is often signalled by the use of wh-questions, rather than intonation on single words. While it is possible to ask a question using only a single word (“tea?”) some of the younger children in our study attempted to complete the task using a question construction such as “would you like some tea?”. In such cases children were reminded to use only a single word but often the metaprosodic leap required to manipulate intonation to do so proved too difficult for young (typically developing) children.

Nevertheless, where floor or ceiling effects are found it is possible that differences are still evident at the brain level. In an fMRI study, Ting Wang et al. (2006) investigated the neural processing of irony in children with ASD. High accuracy levels were reported for both typical children and children with ASD in all conditions, perhaps suggesting the experimental materials used prosody that was unnaturally exaggerated. Despite this,

differences were found at the brain level with the ASD group recruiting prefrontal and temporal regions more strongly overall. Increased task difficulty is associated with greater activation of relevant brain regions, so in ASD more effortful processing was evident. Therefore, where there were high levels of accuracy it is possible that when asked to do this task in real situations the processing would become too effortful and they would fail. Increased activity may reflect compensatory strategies involving the use of verbal reasoning skills for interpreting the communicative intent of others. This resource would not be available in individuals with higher levels of verbal impairment.

2.2.4 *Grammatical Prosody*

Since core linguistic skills are generally not disordered in autism (though they may be delayed) we might expect to find no differences between groups (since they were matched for verbal mental age) in our test of grammatical prosody. This was indeed the case. Again, however, the results suffered from floor effects. Error analyses (paper 3) showed no differences in the comprehension task between groups but in the expression task the children with autism failed to make breaks between items where needed (for example in “chocolate, cake and milk”). Note, however, that in normal conversation such a prosodic minimal pair would be disambiguated by context. Again Table 1 shows a correlation with receptive grammar in the comprehension task, but this time there is no correlation in the expression task. That is, children with autism failed to disambiguate these phrases no matter what their core linguistic skills were.

Since we essentially found floor effects in both children with and without autism (papers 2 and 3), results for grammatical uses of prosody are difficult to interpret. Diehl et al. (2008) looked at the understanding of syntactically ambiguous sentences disambiguated by prosody in 21 adolescents with HFA and 22 typical adolescents matched for IQ and language. They designed their experiment to partial out the effects of syntax and prosody on comprehension, resulting in three conditions:

- Prosody-only: [Put the dog] [in the basket on the star] versus [Put the dog in the basket] [on the star]
- Syntax-only: [Put the dog in the basket that’s on the star] versus [Put the dog that’s in the basket on the star]
- Prosody + syntax: [Put the dog] [in the basket that’s on the star] versus [Put the dog that’s in the basket] [on the star]

Phrases contained either verb phrase (the first of each pair) or noun phrase attachments (the second of each pair). Both groups found the prosody-only condition the most difficult but the HFA group were significantly worse at using prosody alone to disambiguate.

However, in the prosody-only condition, all participants performed better at the verb phrase attachment. This is the high-frequency interpretation, so it is perhaps not surprising that this is the default response (see 2.2.7.2). People with HFA found the syntax-only condition as easy as the syntax + prosody condition suggesting that, unlike typical children, they were not helped by the addition of the prosody. This provides evidence for the idea that the whole prosody system may be impaired, not just paralinguistic prosody even when core linguistic skills are normal, perhaps providing evidence against the dichotomous intonation theory.

Paul et al. (2005a) looked at a wide range of prosodic skills in 27 young people with ASD and 13 controls (not matched). They hypothesised “that the prosodic deficits so frequently attributed to people with autistic syndromes reside primarily in its pragmatic and affective aspects, with grammatical aspects relatively spared” (p206). Therefore, syntactic phrasing, word stress (for example PREsent (noun) versus preSENT (verb)) and turn-end type (questions and statements) are likely to be intact in ASD whereas, affective prosody (emotions: “calm” and “excited” and register: “motherese” and “peer”) and pragmatic prosody (contrastive stress) are likely to be impaired. Results were difficult to interpret due to ceiling effects, but contrary to what was expected, there was a difference in grammatical perception and production of stress and pragmatic perception and production of stress. This suggests a specific difficulty with stress in autism that is not related to the type of prosody, again suggesting that separating prosody into linguistic and paralinguistic types is not particularly useful in autism.

2.2.5 *Prosodic Form*

In PEPS-C (Peppé & McCann, 2003) discrimination tasks are same/different judgements of muffled-sounding versions of real speech materials (actually laryngograph recordings). They therefore contain no lexical information (see appendix one) and are the prosodic equivalent of nonwords. In our studies (papers 2, 3 and 4) children with autism performed more poorly than controls in discrimination, suggesting a basic deficit in auditory perception (which may not be specific to prosody). However, O’Riordan and Passetti (2006) tested the ability of a group of children with autism to discriminate single pure tones and

actually found superior ability. They likened this ability to the well established superiority of discrimination in the visual domain, which is perhaps related to weak central coherence (see 2.2.7.2). Since the stimuli in the O’Riorden and Passetti (2006) study were very short in duration a deficit in auditory memory could explain our differing results. However, Mottron, Peretz and Ménard (2000) tested discrimination of musical melodies of similar duration to our stimuli and also found superior discrimination ability. Like our study, they used a same/different paradigm, but unlike our study, participants had normal language skills and were adolescents. This perhaps suggests that prosody is processed differently from music, however, Järvinen-Pasley and Heaton (2007) tested both discrimination of melodies and similar speech stimuli and found that children with autism showed superior awareness of speech pitch compared to controls. Again, Järvinen-Pasley and Heaton (2007) used a same/different paradigm, which in this case was actually based on the PEPS-C (Peppé & McCann, 2003). However, their study included children with Asperger’s syndrome and all of the children had typical language skills. It is possible that the difficulty our children had with the task was related to their degree of language impairment and indeed there were some correlations with linguistic skills.

Early work by Tallal and Piercy (1973) suggested that SLI, and perhaps other language disorders, may be caused by auditory processing deficits. A review by Rosen (2003) found that auditory deficits are common, if not universal, in SLI. He concludes that the relationship is not causal but there is most certainly an association. It is therefore possible that the children with autism in our studies present with similar problems since they also have co-occurring language disorders which may be similar to SLI (Kjelgaard & Tager-Flusberg, 2001). If this is the case then we would expect children with ASD and no history of language delay, that is children with Asperger’s syndrome, to perform better on the PEPS-C discrimination task. A follow-up study (Peppé et al., in press) on children with Asperger’s syndrome showed this to indeed be the case.

The children with autism were also impaired on their ability to imitate prosody. Hubbard & Trauner (2007) note similar difficulty imitating prosody. In their study they go beyond a subjective rating of the accuracy of an imitation, employing acoustic analysis. They found that in children with autism pitch range was greater upon imitation of affective prosody, suggesting that the participants essentially had exaggerated the intonation they heard. Moreover, they did not use intensity or durational cues reliably, resulting in responses that did not encode the desired emotion accurately.

2.2.6 *Summary of PEPS-C Results*

Papers 2 and 3 highlighted significant differences between children with autism and typical controls in the understanding of affective prosody, and in the ability to express affective prosody and contrastive stress. One major problem with the PEPS-C assessment has been floor effects in tasks leading to non-significant differences between groups of children. Although this makes the results difficult to interpret it is an illustration that prosody continues to develop in the primary school years, with older typically developing children achieving competence in tasks. This highlights the importance of collecting good normative data, and if PEPS-C is to be thought of as a standardised test under development then collecting normative data across a wide age range of children will be essential. Because of this, PEPS-C test results tend to be indicative of the presence or absence of a problem, rather than a full description of the nature of the problem. For example, only one affective distinction is tested, but to truly determine whether a person has disordered affective prosody further testing would be required. Moreover, to determine the nature of an expressive prosodic problem analysis of intonation contours is likely to be necessary. In the clinical context this is not available, but studies are beginning to emerge that look at these specific aspects of prosody in ASDs.

2.2.7 *Explaining Prosodic Impairment: Differences in Cognitive Style*

Although paper 2 showed that prosodic disorder correlated with language disorder, it is unlikely that the relationship is causal since prosodic disorders are more common in ASD than other developmental language disorders and disordered prosody is seen in Asperger's syndrome, an ASD with no language impairment. Moreover, if prosodic disorder was caused by language impairment then we might expect to see more of a deficit in linguistic prosody than paralinguistic prosody, but both types of prosody are impaired. Disordered prosody is a specific feature of autism and we therefore must look to autism-specific explanations at the cognitive level.

Research in ASD is dominated by three main cognitive theories: Theory of Mind (ToM), Weak Central Coherence (WCC) and Executive Dysfunction (EF) (Rajendran & Mitchell, 2007), each discussed in more detail below. ToM has arguably been the most influential of these theories. It is the ability to understand the thoughts, emotions and desires of others (Baron-Cohen, Leslie, & Frith, 1985). WCC (Frith, 1989) is the failure to integrate meaning into higher level representations or difficulty processing information for the whole.

A deficit in central coherence provides advantages too, for example superior performance in tasks which demand attention to high levels of detail. While ToM and WCC are theories driven by what is known about typical cognition, the theory of Executive Dys/function (EF) relies on the behavioural similarity between patients with damage to the frontal lobe and people with autism (Rajendran & Mitchell, 2007). Executive functions include planning, initiation, and inhibition. Historically, the three main cognitive theories have been seen as rivals, with proponents of each theory attempting to explain each aspect of the triad of impairment (see 2). However, attempts to do so have so far been unsuccessful (Happé, Ronald & Plomin, 2006). Happé et al. (2006) propose that just as a single genetic cause of autism is unlikely, a single cognitive explanation is also not plausible and likewise Hill and Frith (2003) suggest that the three theories are not mutually exclusive. It is, however, probable that one theory may have more to offer in explaining prosodic disorders.

2.2.7.1 Theory of Mind

In paper 2 we suggested that the prosodic impairment in autism may be due to a deficit in ToM. Few ToM tasks in the auditory domain exist with the exception of the “Reading the Mind in the Voice task” (Rutherford et al., 2002). In this task listeners are asked to choose a written emotion to match a spoken emotion. Rutherford et al. (2002) suggest that understanding of affect is in itself an advanced test of ToM since the listener must use the speaker’s prosody to infer mental state. In the PEPS-C affect task children were asked to judge whether another person liked or disliked a food-item, based on intonation. It is possible that, due to a deficit in ToM, the children were unable to attribute an emotion to the speaker that was different from their own and therefore approached the task by giving their own preferences.

ToM is known to correlate highly with language skills (Astington & Jenkins, 1999) making it difficult to determine whether a difficulty understanding affect is caused by an early language delay or not. Although prosody correlates highly with language (paper 2) such correlations have not been found in studies of prosody and children with language impairment (Wells & Peppé, 2003) and a recent study of prosody in SLI (using various subtests of the PEPS-C) concluded that prosody was not a core impairment in these children (Marshall et al., 2009). Many studies find a correlational relationship between language and ToM, but how this correlation is interpreted is a matter for much debate in the literature (Siegal & Peterson, 2008). A study by de Villiers and Pyers (2002) suggested that children need the complex syntax of mental verbs and an understanding of sentential complements (such as “Sarah thought the Earth was flat”) in order to represent the beliefs of others. In

other words, a correlation between language skills as measured by a standardised language test and affective prosody (that is, the ability to understand another's ToM based on emotion in their voice) may be due to increased scores on the language test due to children having acquired the specific grammatical structures noted by de Villiers and Pyers (2002). This would explain the correlation we found between the PEPS-C affect input task and two out of three of our language measures.

However, Siegal and Peterson (2008) refute this claim, citing evidence from studies of young children with good syntactic skills and yet poor ToM. They suggest that impaired ToM is the result of reduced exposure to social communication at a young age. Evidence from studies of deaf children not exposed to native signers or blind children supports this, since children with sensory impairments are less likely to engage in social exchanges with parents and peers at a young age. In the same way, children with autism, perhaps due to auditory processing deficits, are less likely to attend to social exchanges at a young age and therefore develop deficits in ToM and language (Siegal & Peterson, 2008).

If we think of affective prosody as a ToM task in itself, then our finding of no correlation between the ability to convey different types of prosodic affect and language may be further evidence for the dissociation suggested by Siegal and Peterson (2008). Since the children with autism cannot theorise that others have thoughts different from their own they may not find it necessary to use a device such as prosody to enhance meaning, since they may believe that the listener will have the same opinion as them.

Similarly, impaired ToM may explain a difficulty with pragmatic uses of prosody such as contrastive stress. ToM is implicated in explanations of how speakers choose referential expressions (Arnold, Bennetto & Diehl, 2009) since speakers only use underspecified expressions such as pronouns when they can assume that the referent is known to the listener. This therefore requires the speaker to have a theory of mind. It is possible that contrastive stress requires the same kind of knowledge since emphasising a word in an utterance requires the speaker to have understood what the listener's prior knowledge is likely to be. People with autism may have difficulty selecting which word in an utterance should be in focus since they lack the ToM required to do this.

It is more difficult to explain Paul et al. (2005) and Diehl et al.'s (2008) findings of impairment in grammatical prosody using the ToM account. It is plausible that prosody is generally impaired because people with autism do not realise that prosodic information is useful, if not usually essential, to the listener. In everyday conversation prosody generally

enhances the meaning of an utterance, rather than changes it (except in perhaps irony). However, in the PEPS-C tasks an understanding of prosody is crucial to success.

2.2.7.2 *Weak Central Coherence*

In paper 3 we suggest that WCC may be a candidate for explaining prosodic impairment in autism. WCC implies a preference for local over global processing and has been found repeatedly in autism, with most experiments investigating the visual domain. People with autism are known not to succumb to visual illusions (Happé 1996), perhaps because they fail to integrate all of the visual information into the gestalt. Similarly, it is possible that people with autism would not succumb to misleading prosody (an “auditory illusion”) because of a difficulty integrating all the strands of information in the acoustic signal. For example, in irony typical people are more persuaded by prosody than the literal meaning of an utterance but people with ASD tend to interpret irony literally (Happé, 1995). This has been said to be due to poor ToM, but studies have not taken prosody into account and any difficulty in this area may be due to a difficulty integrating information. As prosody often enhances rather than changes the meaning of an utterance; this may lead to it being treated as non-essential. Lexis and syntax are, on the other hand, always essential. It is therefore plausible that where there are competing strands of information prosody may go unprocessed, or be perceived as unimportant, with lexis and syntax taking preference.

For example, studies of processing homographs in context (for example López & Leekam, 2003) show that children with autism always opt for the high frequency, marked, pronunciation of homographs. Compare this with Paul et al.’s (2005) lexical stress task (PREsent (noun) versus preSENT (verb)) and it is easy to see how the individuals with autism may have difficulty with this task. Similarly, in the Diehl et al. (2008, see 2.2.4) study the high-frequency interpretation was the default response. The problem is further compounded by a difficulty integrating facial expression (so called, “visual prosody”) with auditory cues (Swerts, 2009), again highlighting a difficulty with central coherence.

2.2.7.3 *Executive Dysfunction*

Both ToM and WCC can be used as cognitive explanations of deficits in prosody, but it is more difficult to explain a prosodic deficit using the executive dysfunction account. One possible explanation is that people with autism misunderstand prosody that is at odds with the literal meaning of an utterance because they find it difficult to inhibit the default, unmarked, response. For example, in sarcasm it is possible that people with autism fail to use prosody to interpret the utterance correctly because a deficit in EF means that they cannot

inhibit the literal response. Few studies link the communication deficits in autism to executive dysfunction. Joseph, McGrath and Tager-Flusberg (2005) found that although children with autism had deficits in both EF and language there was no direct relationship between the two.

2.3 Speech and Prosody in ASD: Summary

Paper 1 demonstrated that prosody in autism was an under-researched area. This is surprising since unusual expressive prosody was noted in Kanner's original description of autism (Kanner, 1943). Coupled with residual articulation disorders (2.1, Gibbon et al., 2004; Shriberg et al., 2001 and Cleland, Gibbon et al., 2010), this adds a major communication barrier for people with ASD. Paper 3 highlighted that prosody is impaired in both the expressive and receptive domain and paper 2 suggests that degree of prosodic disorder is highly correlated with language ability. However, paper 4 illustrates that in individual children with autism it is possible to present with prosodic impairment and normal language skills, suggesting dissociation. A detailed analysis of intonation patterns was beyond the scope of our work; however, paper 1 shows this to be an area for further development. Some recent research (Green & Tobin, 2009) using acoustic analysis of intonation patterns in Israeli Hebrew shows that children with ASD are able to produce a wide variety of prosodic patterns but choose only to use a limited repertoire. Whether this is a conscious decision is not clear, but the idea that speakers with autism do not provide their conversational partners with as much information as possible to help interpret their message is consistent with a deficit in ToM. A deficit in ToM explains disordered affective, and perhaps pragmatic, prosody but does not explain disorders in grammatical prosody as easily. Future studies of prosody in autism should look also to WCC as a failure to integrate information from multiple contexts seems a likely explanation for the difficulties.

3 Down's Syndrome

In our research (papers 5 and 6) we sought to establish whether severity of speech disorder correlated with severity of linguistic or cognitive delay. While disordered prosody is a frequent characteristic of autism, articulation and phonology are specifically and severely impaired in DS. Both developmental disorders therefore feature speech that is different to the norm, making people stand out as different to their peers. In autism we found that severity of prosodic disorder correlated with severity of language impairment, but minor speech disorders did not.

In our review of the literature on speech in DS (Timmins, Cleland, Rodger, et al., 2009) we found evidence of impairments in almost all of the mechanisms required for successful speech but little consensus on what is the primary speech disorder. Despite this, most previous studies have taken a purely phonological approach (e.g. Stoel-Gammon, 1980; Van Borsel, 1996), implying that the speech disorder is purely linguistic in nature. However, because DS includes an anatomical and physiological profile that could lead to certain types of speech errors (for example, hypotonia may lead to dysarthria which in turn causes weak articulations and distortions), in our studies (papers 5 and 6) we do not take the purely phonological approach of previous studies. Instead we described all of the errors, both phonetic and phonological, produced by children and young people with DS. By using fine phonetic transcription and instrumental techniques we were able to determine whether children with DS produced speech errors that are not found in typical development. This type of analysis allows differential diagnosis of the speech disorder in DS beyond a phonological account.

3.1 Speech in DS: Error Types

Stoel-Gammon (2001) lists the phonological processes seen in children with DS as cluster reduction, final consonant deletion, stopping, prevocalic voicing, gliding, vocalisation and final consonant devoicing: in other words, common developmental processes. In our study of speech errors in DS (paper 5) we found all of these errors. Speech disorders ranged from mild to severe and a few children were excluded from the study because their phonetic inventory included no consonants (these children were using alternative communication such as sign), suggesting a profound speech impairment. Like previous studies, delayed phonological processes were common. Cluster reduction was the most common process, displayed by all of the children bar one. Other structural simplifications were also common, final consonant deletion being displayed by all of the children. These results suggest, like previous literature, a pattern of phonological delay. However, the non-developmental process of initial consonant deletion was also seen in the majority of children. Together the structural processes were more common than the systemic ones, suggesting that most children with DS use reduced forms. In some children omissions affected nearly every consonant in the word, leaving only the vowel. Rupela and Manjula (2007) also describe phonotactic patterns in DS (in the language Kannada). They compared speakers with DS to speakers with other learning disabilities and found that people with DS used simpler phonotactic patterns. A large number of omissions may be associated with dyspraxia, rather than delayed phonology and given

that the children also omitted word initial consonants this diagnosis should be investigated further (see 3.3).

Figure 2 (adapted from paper 5) shows all the processes displayed by the children with DS with developmental processes in white and non-developmental processes in black.

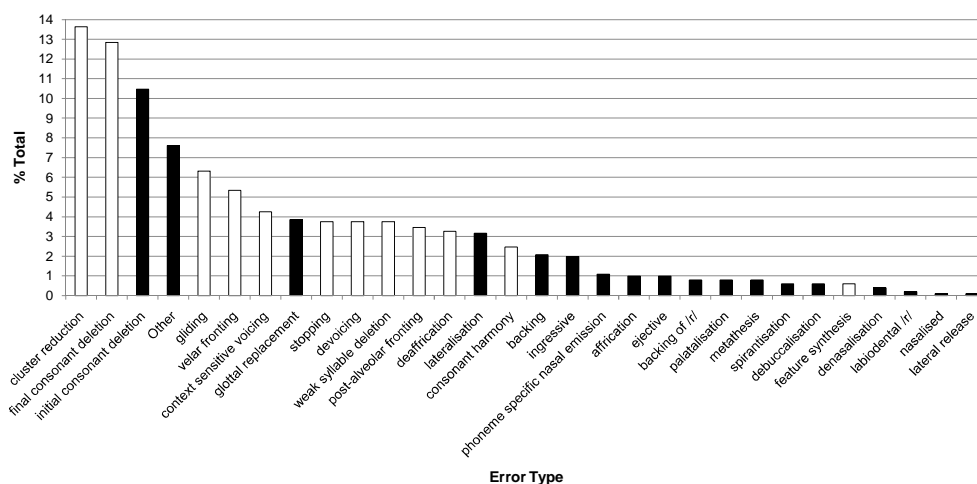


Figure 2: Relative occurrence of a range of speech error types in children with DS (Developmental errors are shown in white and non-developmental errors in black).

Twenty-nine different error types were described in the children with DS, and while developmental processes were more prevalent there were more categories of non-developmental or unusual errors. Many of the errors have not been reported before in the speech of children with DS. For example, one child's speech was characterised by pulmonic ingressive fricatives. Non-pulmonic airstream consonants were also found with ejectives being common, though note that ejectives may not always be disordered (Scobbie, Gordeeva & Matthews, 2006). We conclude that speech in DS is characterised by atypical errors alongside many developmental errors and increased omissions are very common. In children with speech impairments who are otherwise typically developing, this would usually constitute a disorder rather than a delay.

Most studies presume that since the majority of errors can be classified as known developmental processes that the cause is a delay in the phonological system. This conclusion is problematic since most studies use broad transcription and do not investigate oromotor skills even though these are known to be impaired (Barnes et al., 2006). In paper 5

children presented with reduced oromotor function as expected and this correlated with percentage phonemes correct. If errors were caused solely by phonological impairments we would not expect to see such a correlation.

Despite the well-known anatomical and physiological problems in DS, few studies report the distortions that we might expect. In paper 6 we looked at fewer (six) children in more detail and reported several types of distortions. These distortions mainly affected sibilants which were realised as laterals, or central+laterals. Van Borsel (1996) reported similar distortions. He compared a group of young people with DS (aged 15;4-28;3) to a control group of children young enough still to be in the process of phonological acquisition (aged 2;6-3;4). Since many of the speech errors were similar between the two groups he concluded that phonology was delayed in DS. However, he reported additional distortions in the DS group, for example, “denasalisation”, “dentalisation” and “wet”. This finding is largely ignored in the Van Borsel study, with the author choosing to focus on delayed phonology as a cause of reduced intelligibility in DS. This conclusion is problematic since distortions such as these are phonetic rather than phonological, perhaps caused by anatomical differences or dysarthria.

Phonetic distortions tend not to be recorded when using broad transcription alone. Studies which use only broad transcription tend therefore to report mainly delayed phonological processes, giving the impression that the speech of people with DS will sound very much like that of younger children. In fact it is much less intelligible than the reported delayed phonological processes would imply. In paper 6 we use both fine phonetic transcription and the instrumental technique electropalatography (EPG, Hardcastle, 1972), to allow us to identify subtle differences in linguapalatal contact patterns between children with DS and typical children. EPG reveals patterns that differ from typically developing children (Timmins, McCann, Wood et al., 2009) and are disordered in nature, even when perceptually correct.

By looking at speech in this way we identified errors that may previously have been described as delayed phonological processes but were actually far more complex. For example, in paper 6, Child 5 presented with an atypical case of velar fronting. Most (60%) productions of velars were realised as alveolars and as such would be classified as fronted, due to the delayed resolution of a developmental phonological process. However, EPG revealed that some of these attempts were double articulations (simultaneous alveolar and velar closure) and attempts that were transcribed as [t] were abnormally retracted. Since this child was producing quantifiable, if not audible, differences between /t/ and /k/ (i.e. a covert

contrast, Scobbie et al., 1997) then it is unlikely that the problem is at the phonological level. However, since EPG is not routinely used with children who have phonological delay we cannot be sure that these errors are not seen as part of what has previously been described as velar fronting, but this is a far wider problem than can be addressed here. Overall, there were a large number of disordered patterns and distortions, suggesting that the speech disorder in DS goes beyond a delay in phonological acquisition.

3.2 *The Relationship between Speech, Language and Cognition*

All individuals with DS present with some degree of intellectual impairment (Roizen, 2002). However, speech is particularly impaired and since cognition is delayed it is tempting to conclude that speech delay would be commensurate with cognitive skills. If, as previous studies have implied, speech delay/disorder is related to a general delay in cognition and language we might expect to find correlations between these measures. In paper 5 we show this not to be the case. In fact some children with the most severe speech disorders had above average (compared to other children with DS) levels of cognitive ability and vice versa. However, like children with ASD (see 2.1) it may still be possible that a cognitive or language delay is responsible, at least in part, for a delay in acquisition.

It is conceivable that developmental processes might correlate with language or cognition but non-developmental processes might not. A closer look at the data in paper 5 shows this is not so, with no correlations between the number of developmental errors and cognitive or language measures or between atypical errors and these measures. This is not surprising given that in children with language impairment co-morbid speech disorders are not that common. In fact Shriberg, Tomblin and McSweeney (1999) reported the co-morbidity of speech delay with cognition/language impairment to be less than 2%. The evidence suggests that the speech disorder in DS is not a direct result of cognitive or language impairment. Further evidence for this comes from comparisons between DS and other learning disabilities. When children with DS are cognitively matched to children with other intellectual impairments such as Fragile X, speech is always more impaired in DS (for example Abbeduto et al., 2001 and Barnes et al., 2006). Clinically this finding is crucial. In some speech and language therapy services priority is based on discrepancy criteria; where if a speech or language skill is found to be in line with cognitive level then direct therapy is not provided. In DS this is not the case: speech is specifically and often severely impaired and therefore warrants consideration for treatment.

Despite this, treatment studies are few. This is surprising, given both our findings and the fact that DS is an easily identifiable and common condition. In paper 6 we found that children and young people with DS responded well to both visual feedback therapy using EPG therapy (Hardcastle & Gibbon, 1997) and to other types of speech therapies such as Core Vocabulary therapy (Dodd et al., 2006) suggesting that speech-focused intervention is useful.

3.3 Differential Diagnosis of Speech Disorders in Down's Syndrome

Our studies provide clear evidence that the speech disorder in DS is not simply a delay in phonological acquisition. However, for clinicians to provide effective therapy they need to determine the primary diagnosis. Even if children show distortions and subtle phonetic differences in their speech, perhaps due to anatomical differences, this does not necessarily preclude a delay in phonology as a primary diagnosis, suggesting that phonological therapy may be effective. Since hypotonia is involved, dysarthria is also a candidate for a primary diagnosis, and likewise children with DS show characteristics of dyspraxia (Kumin, 2006).

Most studies of speech in DS subscribe to the notion that phonology is delayed, or delayed with elements of disorder, without considering that some of the processes described could be the result of a motor or anatomical impairment. For example child 5 (see 3.1) in paper 6 could be described as velar fronting but EPG analysis showed his errors to be more phonetic in nature. For this particular child a diagnosis of dyspraxia was suggested since his productions were highly inconsistent and he (like most people with DS) showed impaired oromotor skills (Dodd et al., 2002). While numerous studies have sought to characterise dyspraxia, most exclude children with cognitive impairments or obvious muscle weakness. This has led to people with DS not usually being considered for a diagnosis of dyspraxia, moreover, due to the presence of hypotonia treatment is usually undertaken from a dysarthria point of view (Kumin & Adams, 2000). However, in paper 5 we suggest that children with DS show many of the characteristics of dyspraxia such as processes that are hard to classify, limited phonetic inventory (ASHA, 2007) and omission of sounds and syllables (Rupela & Manjula, 2007).

In order to determine whether children with DS present with more of a dysarthric or dyspraxic profile a new diadochokinetic (DDK) task specifically for children with cognitive impairments has been designed (McCann & Wrench, 2007). Although maximum performance tasks such as DDK assess abilities that differ from those used in typical speech

production, they can provide information on the motor speech impairments that underlie dysarthria and dyspraxia (Thoonen et al., 1996). Oromotor DDK assesses performance in rapidly alternating movements (Fletcher, 1978), usually repetition of syllables, [tə tə tə], or sequences of syllables, [pə tə kə], at maximum rate. In typical development, DDK rates increase with age (Fletcher, 1978) and slow DDK rates may be indicative of speech disorders (Williams & Stackhouse, 2000). Thoonen et al. (1996) suggest that children with dysarthria produce significantly slower monosyllabic repetitions rates than both children with dyspraxia and typical children. In contrast, children with dyspraxia do not show reduced rates for monosyllables but do show some reduction in the trisyllabic condition. Most importantly, they showed serious inaccuracy in the trisyllabic condition. In McCann and Wrench (2007) we found that a small group of children with DS produced similar DDK rates to typically developing children. By comparing data from a larger group of 21 children with DS (aged 9;2 to 18;9, mean=13.47, SD=3.15) to published norms (St Louis & Ruscello, 1987) we can determine whether DDK rates are within the normal range. Since disorder is usually defined as at least one standard deviation below the normal range we can compare our group of children with DS to the mean \pm 1SD for adolescents (aged 19;8: this gives a conservative estimate). Figure 3 shows the DDK rate for both monosyllables (a mean of [pə], [tə] and [kə] repetition at maximum rate, see McCann & Wrench, 2007) and trisyllables (maximum rate of [pə tə kə], see McCann & Wrench, 2007). The mean \pm 1 S.D. from St Louis and Ruscello (1987) is denoted by shading ($M=5.05$, $SD=.65$), showing that most of the children (81%) have DDK rates within or above the normal range, suggesting that for most dysarthria is perhaps not the primary diagnosis. However, all of the children had (uncorrelated) trisyllabic rates below the normal rate (below $6.67 \pm .96$), which is indeed indicative of dyspraxia.

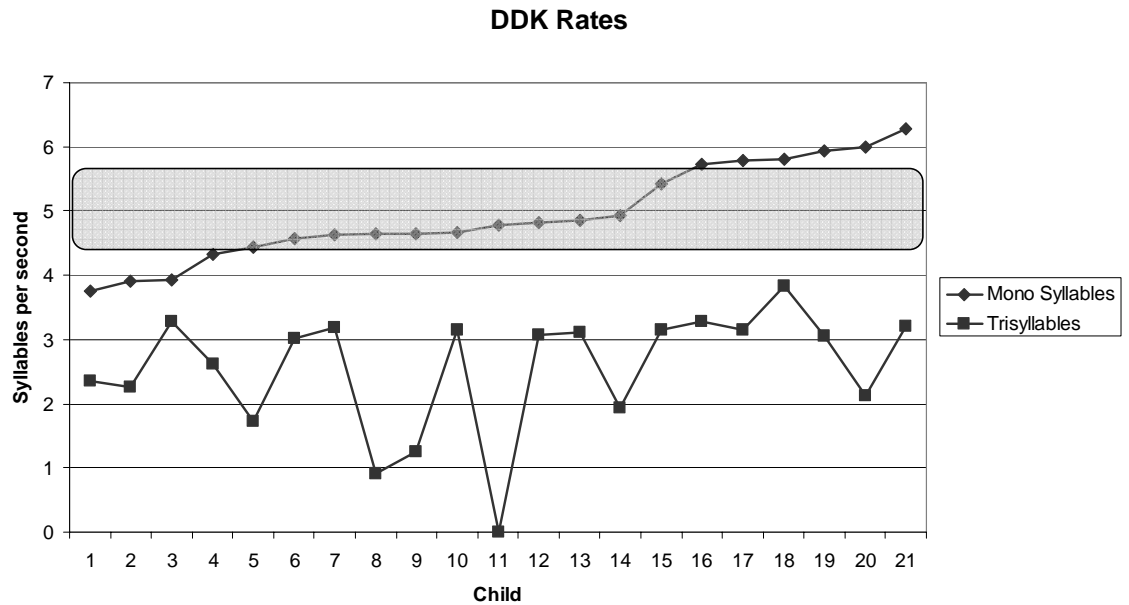


Figure 3: DDK rates (syllables per second) for children with DS. Shading denotes normal range for maximum repetition of monosyllables.

Accuracy of consonant production is ignored in many DDK studies. However, difficulty in sequencing is one of the diagnostic features of dyspraxia and therefore inaccuracy in the tri-syllabic condition may be diagnostic of dyspraxia. In McCann and Wrench (2007) we show that accuracy is seriously impaired in children with DS, with most children unable to sequence /ptk/ at all. This provides good evidence that dyspraxia should seriously be considered as a primary diagnosis for children with DS, despite their obvious muscle weakness. Kumin and Adams (2000) suggest that because children with DS are known to have sensory and motor deficits this would lead to an impaired ability at the cognitive level to form motor plans (templates) for speech, leading to speech becoming a conscious effort rather than an automatic process. This leads to behavioural features of dyspraxia such as decreasing intelligibility with increasing utterance length, difficulty sequencing and groping.

3.4 *Speech in DS: Summary*

Previous research has suggested that the speech of people with DS is characterised by delayed phonology but in paper 5 we showed that all participants with DS also presented with disordered productions. By using fine phonetic transcription we were able to describe the types of errors seen in children and adolescents with DS, but determining the cause of the

errors required instrumental analysis (paper 6) and oromotor assessment (McCann & Wrench, 2007 and 3.3). EPG analysis in paper 6 showed that children with DS were producing speech that was different from typical children in subtle ways not revealed by broad transcription. Moreover, errors which may be mistakenly attributed to delayed phonological processes (paper 6, child 5, see 3.1) may in fact be covertly contrastive, suggesting a phonetic explanation. Where subtle distortions are found, the cause may be anatomical but there is also good evidence to suggest that at least some children with DS present with dyspraxia.

When taken together this evidence suggests that clinicians need to look beyond a narrow linguistic-phonological account of speech disorders in DS, applying their differential diagnosis skills to each child in order to plan appropriate interventions. Moreover, since the speech disorders in DS are not related to a general delay in language or cognition (paper 5) and are in fact usually much more severe, speech-focussed intervention is particularly warranted.

4 Summary and Conclusions

Previous studies of developmental language disorders focused their efforts on three areas (Rice, Warren & Betz, 2005): Describing the language and cognitive profile of disorders; determining whether language and cognition is delayed or disordered and determining whether language skills are related to, or correlate with, nonverbal skills. The programme of research described here adds to this discussion by addressing speech and prosody, and by relating each of these domains to language and cognition. Autism and Down's syndrome were chosen as the two developmental disorders that present with specific deficits in prosody (ASD, papers 1, 2, 3 and 4) and speech (DS, papers 5 and 6).

In both autism and DS we see delayed language, especially expressive language. Although speech was much more severely impaired in DS than it was in ASD, both conditions presented with errors that could be categorised as delayed phonological processes. However, our results show that both conditions also present with distortions that are more phonetic in nature and with errors that do not appear in typical development such as phoneme specific nasal emission and ingressive fricatives. These findings suggest that minor delays and distortions persist in ASD due to a lack of ability to identify with peers whereas in DS anatomical differences and difficulty with motor planning are the likely causes. For both conditions speech and/or prosody are unusual sounding at a phonetic level in addition to being systematically immature.

For autism, the problem of unusual sounding speech is compounded by receptive and expressive prosodic disorder. Despite disordered prosody being a specific feature of ASD very little previous research has addressed the nature of this disorder. Early studies focused on stress (see paper 1), finding it to be atypical, but few studies looked at the understanding of prosody. Prosody in children with ASD differed from that of typical children, indicating disorder rather than delay. Like studies of visual processing of emotion, affective prosody was particularly impaired, as was contrastive stress. Children with autism had serious difficulty with the form tasks in our studies (papers 2 and 3). This suggests that there is some difficulty with discriminating prosody at a basic level and with using prosodic forms. Although we did not find difficulties with grammatical prosody in our studies this was mainly due to floor effects. Other authors (Diehl et al., 2008 and Paul et al., 2005) have found difficulties in this area. Taken together these findings suggest that prosody is impaired across the board in autism, not just in affective and pragmatic, or paralinguistic, uses. This is evidence against both the dichotomous intonation theory and the notion that disordered prosody is the result of language impairment in autism. Disordered prosody seems to be a specific behavioural feature of autism and as such cognitive models of autism must take it into account. Impaired theory of mind can explain those difficulties associated with affective prosody, however, weak central coherence has more to offer the study of prosody in autism generally. If people with autism have difficulty integrating multiple strands of information, it is possible that where there are competing strands of information prosody may be likely to go unprocessed, or perceived as unimportant, in preference of other sources of information such as lexis and syntax.

In order to determine whether speech and prosody development is independent of general cognitive and language development, papers 2 and 5 focused specifically on the relationships between speech, prosody and formal linguistic and nonverbal skills. For both DS and ASD we found no relationship between speech (percentage consonants correct) and either language or nonverbal ability. Despite the presentation and causes of speech disorders in DS and ASD being so different, especially in terms of severity, it appears that disorder in this area is relatively independent of nonverbal cognition and language (or at least grammar and vocabulary as measured by standardised assessments). Speech in DS is severely and specifically impaired with phonetic distortions that may be due to anatomical differences and problems with motor programming. This has important clinical implications because it suggests that speech intervention may be warranted. Choosing the type of intervention is not straightforward. Although both groups presented with delayed phonological processes, phonological therapy approaches may not be suitable with our DDK assessment (3.3)

showing that dyspraxia is likely to be the primary cause of reduced intelligibility in DS. In order to plan appropriate interventions clinicians should apply their skills in differential diagnosis to each client, regardless of their developmental disorder.

It is clearly illustrated here that the use of standardised articulation and phonology tests is problematic, especially when standard scores from articulation tests are compared to other language and cognitive tests (see 2.1). The same cautions apply to prosodic assessment. Few tests of prosody exist and none are yet standardised. It is not yet clear whether prosody is normally distributed in the school-aged population, and our research suffered from floor effects (papers 2 and 3), suggesting that looking at older children or children without language impairments may be a useful approach. Nevertheless, compared to assessment of other aspects of speech and language, prosody assessment is in its infancy (Diehl & Paul, 2009) and we should therefore use our knowledge of assessment of segmental disorders to inform our assessment of suprasegmental disorders, working towards developing a standardised test of prosody. It is also important to remember that the subtests in the PEPS-C are not of equal difficulty, children acquire different prosodic skills at different ages. However, this issue is also found in phonological assessment, overall scores may be used to judge whether an impairment exists, but since different phonemes are acquired at different ages an error analysis is always required before the clinician can determine both the significance and nature of the impairment. The same must be true for prosodic assessment. It is important that any standardisation of a prosodic assessment such as PEPS-C is not at the expense of analysis of comprehension and expression of different prosodic functions.

Furthermore, while the function tasks in the PEPS-C tell us whether a person with ASD is likely to have difficulty expressing different functions of prosody they do not tell us what a person with ASD is likely to sound like. We have shown here the importance of describing phonetic distortions when dealing with segments (see 2.1 and 3.1) and again prosodic disorders ought to require similar analyses since, for example, monotonous or singsong intonation sound very different but may result in the same impairments in function. The expressive form tasks go some way to addressing this, but simply identify the presence of a problem, rather than describing it. This is a similar problem to the use of right/wrong scoring on an articulation test which we overcame using fine phonetic transcription and instrumental techniques. For speech, using these techniques has shown us that articulation is not spared in autism as had previously been thought (Kjelgaard & Tager-Flusberg, 2001) and that errors in both DS and ASD are not simply delayed phonological processes but are in fact disordered and may be phonetic in nature. Future research in expressive prosody in developmental disorders needs to look more at these issues, using both transcription (Müller

& Ball, 2009) and acoustic analysis to identify differences in intonation between speakers with ASD and typical speakers.

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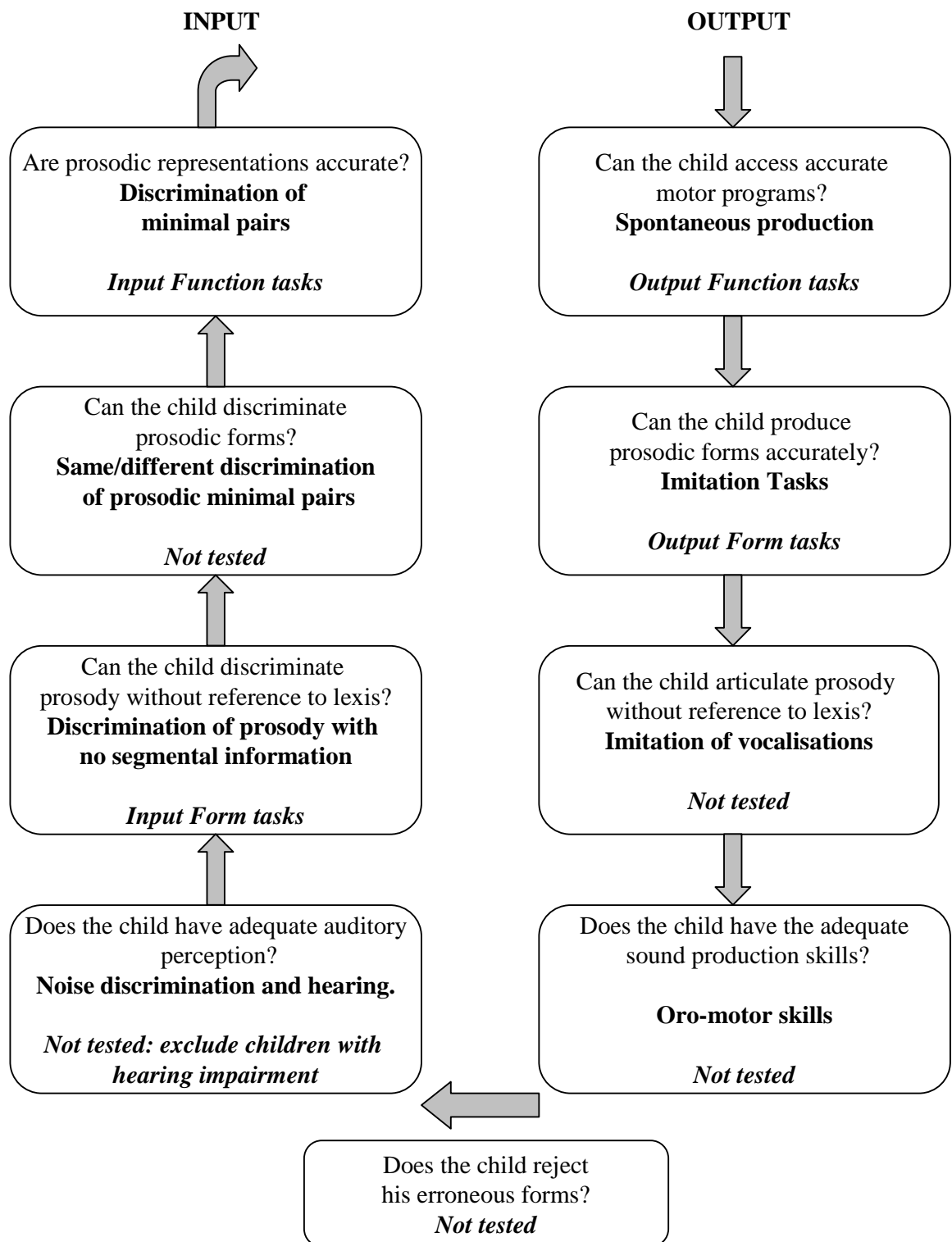
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Appendix One: Prosodic Processing Model



Stackhouse and Wells (1997) psycholinguistic framework, modified to represent the stages required for successful perception and production of prosody. PEPS-C tasks are in *italics*.

Appendix Two: Abstracts from Selected Publications.

Paper 1

McCann, J. & Peppé, S. (2003). Prosody in Autism Spectrum Disorders: A Critical Review. *International Journal of Language and Communication Disorders*, 38, 325-350.

<http://informahealthcare.com/doi/pdf/10.1080/1368282031000154204>

Background: Many individuals with autism spectrum disorders present with unusual or odd-sounding prosody. Despite this widely noted observation, prosodic ability in autism spectrum disorders is often perceived as an under researched area.

Aims: This review seeks to establish whether there is a prosodic disorder in autism, what generalizations can be made about its various manifestations and whether these manifestations vary according to the diagnosis. A literature review was carried out to establish what areas of prosody in autism spectrum disorders have been researched to date, what the findings have been and to determine what areas are yet to be researched.

Main contribution: It is shown that prosody in autism spectrum disorders is an under-researched area and that where research has been undertaken, findings often conflict. The findings of these conflicting studies are compared and recommendations are made for areas of future research.

Conclusions: Research in this area has covered mostly prosodic expression, although some more recent studies cover comprehension, processing and the relationship of receptive prosodic ability to theory of mind. Findings conflict and methodology varies greatly.

McCann, J., Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Prosody and its relationship to language in school-aged children with high-functioning autism. *International Journal of Language and Communication Disorders*, 42, 682-702.

<http://dx.doi.org/10.1080/13682820601170102>

Background: Disordered expressive prosody is a widely reported characteristic of individuals with autism. Despite this, it has received little attention in the literature and the few studies that have addressed it have not described its relationship to other aspects of communication.

Aims: To determine the nature and relationship of expressive and receptive language, phonology, pragmatics, and non-verbal ability in school-aged children with high-functioning autism and to determine how prosody relates to these abilities and which aspects of prosody are most affected.

Methods & Procedures: A total of 31 children with high-functioning autism and 72 typically developing children matched for verbal mental age completed a battery of speech, language, and non-verbal assessments and a procedure for assessing receptive and expressive prosody.

Outcomes & Results: Language skills varied, but the majority of children with high-functioning autism had deficits in at least one aspect of language with expressive language most severely impaired. All of the children with high-functioning autism had difficulty with at least one aspect of prosody and prosodic ability correlated highly with expressive and receptive language. The children with high-functioning autism showed significantly poorer prosodic skills than the control group, even after adjusting for verbal mental age.

Conclusions: Investigating prosody and its relationship to language in autism is clinically important because expressive prosodic disorders add an additional social and communication barrier for these children and problems are often lifelong even when other areas of language improve. Furthermore, a receptive prosodic impairment may have implications not only for understanding the many functions of prosody but also for general language comprehension.

Peppé, S., McCann, J., Gibbon, F. & O'Hare, A. (2007). Receptive and expressive prosodic ability in children with high-functioning autism. *Journal of Speech, Language, and Hearing Research*, 50, 1015-1028.

<http://jslhr.highwire.org/cgi/reprint/50/4/1015>

Purpose: This study aimed to identify the nature and extent of receptive and expressive prosodic deficits in children with high-functioning autism (HFA).

Method: Thirty-one children with HFA, 72 typically developing controls matched on verbal mental age, and 33 adults with normal speech completed the prosody assessment procedure, Profiling Elements of Prosodic Systems in Children.

Results: Children with HFA performed significantly less well than controls on 11 of 12 prosody tasks ($p < .005$). Receptive prosodic skills showed a strong correlation ($p < .01$) with verbal mental age in both groups, and to a lesser extent with expressive prosodic skills. Receptive prosodic scores also correlated with expressive prosody scores, particularly in grammatical prosodic functions. Prosodic development in the HFA group appeared to be delayed in many aspects of prosody and deviant in some. Adults showed near-ceiling scores in all tasks.

Conclusions: The study demonstrates that receptive and expressive prosodic skills are closely associated in HFA. Receptive prosodic skills would be an appropriate focus for clinical intervention, and further investigation of prosody and the relationship between prosody and social skills is warranted.

McCann, J., Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2008). The Prosody-Language Relationship in Children with High-Functioning Autism. In *McGregor, E., Nunez, M., Cebula, K. and Gomez, J.C. (Eds.) Autism. An Integrated View from Neurocognitive, Clinical, and Intervention Research.*, Oxford, United Kingdom, Blackwell, 214-235.

<http://eresearch.qmu.ac.uk/1544/>

Introduction: Kanner included unusual prosody as part of his original description of autism in 1943: many of the children spoke in a monotonous, abrupt or singsong way, or with a voice “peculiarly unmodulated, somewhat hoarse” (p.241). A simple definition of prosody is that it refers to the manner in which things are said, not the content of what is said. The manner is conveyed by a number of different factors: variations in the relative pitch and duration of syllables, loudness of voice, pauses, intonation, speech-rate, stress and speech-rhythm. Disordered expressive prosody is widely reported to occur in the speech of people with autism (for example, Baltaxe, 1984; Fine, Bartolucci, Ginsberg, & Szatmari, 1991; Shriberg, Paul, McSweeney, Klin, Cohen, & Volkmar, 2001) but very little empirical or clinical research has been conducted on this aspect of autism. A recent review (McCann & Peppé, 2003) found only 16 studies between 1980 and 2002 on the topic. Of these, only two considered receptive prosodic disorder, which may not only account, at least in part, for expressive disorder, but also be related to the language disorders so frequently seen in autism. This chapter provides an overview of prosodic skills in autism and how these may link with language development more broadly. It then goes on to describe our recent research, which aimed to develop methods of prosodic assessment and provide finer-grained information on the links with aspects of language development. Two case-studies are included to further illustrate this relationship.

Cleland, J., Wood, S., Hardcastle, W., Wishart, J. & Timmins, C. (2010). The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. *International Journal of Language and Communication Disorders*, 45(1), 83-95.
<http://dx.doi.org/10.3109/13682820902745453>

Background: Children and young people with Down's syndrome present with deficits in expressive speech and language, accompanied by strengths in vocabulary comprehension compared with non-verbal mental age. Intelligibility is particularly low, but whether speech is delayed or disordered is a controversial topic. Most studies suggest a delay, but no studies explore the relationship between cognitive or language skills and intelligibility.

Aims: This study sought to determine whether severity of speech disorder correlates with language and cognitive level and to classify the types of errors, developmental or non-developmental, that occur in the speech of children and adolescents with Down's syndrome.

Methods & Procedures: Fifteen children and adolescents with Down's syndrome (aged 9–18 years) were recruited. Participants completed a battery of standardized speech, language and cognitive assessments. The phonology assessment was subject to phonological and phonetic analyses. Results from each test were correlated to determine relationships.

Outcome & Results: Individuals with Down's syndrome present with deficits in receptive and expressive language that are not wholly accounted for by their cognitive delay. Receptive vocabulary is a strength in comparison with expressive and receptive language skills, but it was unclear from the findings whether it is more advanced compared with non-verbal cognitive skills. The majority of speech errors were developmental in nature, but all of the children with Down's syndrome showed at least one atypical or non-developmental speech error.

Conclusions & Implications: Children with Down's syndrome present with speech disorders characterized by atypical, and often unusual, errors alongside many developmental errors. A lack of correlation between speech and cognition or language measures suggests that the speech disorder in Down's syndrome is not simply due to cognitive delay. Better

differential diagnosis of speech disorders in Down's syndrome is required, allowing interventions to target the specific disorder in each individual.

Cleland, J., Timmins, C., Wood, S., Hardcastle, W. & Wishart, J. (2009). Electropalatographic therapy for children and young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23(12), 926-939.

<http://dx.doi.org/10.3109/02699200903061776>

Articulation disorders in Down's syndrome (DS) are prevalent and often intractable. Individuals with DS generally prefer visual to auditory methods of learning and may therefore find it beneficial to be given a visual model during speech intervention, such as that provided by electropalatography (EPG). In this study, participants with Down's syndrome, aged 10:1 to 18:9, received 24 individualized therapy sessions using EPG. Simultaneous acoustic and EPG recordings were made pre- and post-intervention during 10 repetitions of a word list containing lingua-palatal consonants. Participants also completed the DEAP phonology sub-test at both time points. Post-treatment, all participants showed qualitative and quantifiable differences in EPG patterns and improvements in DEAP percentage consonants correct. EPG assessment and therapy appears a positive approach for identifying and improving articulatory patterns in children with DS.

Appendix Three: Co-Author Statements

Autism Papers

Statement from Sue Peppé on behalf of all co-authors.

Joanne was Research Assistant on two projects for which I was the Project Manager. The first project, entitled ‘Prosodic ability of children with high-functioning autism’ ran from May 2002-April 2004 (funded by the Scottish Health Executive’s Chief Scientist Office, £135,438). The second, ‘Prosodic ability of children with Asperger’s syndrome’, ran from May 2004-September 2006 (funded by the ESRC, £189,924).

Joanne’s role included the following:

- researching the background literature for the projects
- helping to design tasks to assess prosody skills: researching stimuli, and difficulty levels, recording and editing stimuli;
- writing the instructions to be implemented in the computer program for the tasks, liaising with computer consultants
- organising and maintaining contacts with participants, liaising with schools and parents
- interviewing participants (70 with autistic spectrum disorders, 70 typically-developing children)
- keeping data records
- writing up papers reflecting findings
- presenting project data

1. **McCann, J. & Peppé, S. (2003)** Prosody in Autism Spectrum Disorders: A Critical Review. *International Journal of Language and Communication Disorders*, 38(4), 325-350.

This paper reviews the literature relating to prosody skills in autism since 1980. Joanne had researched much of this literature as part of her honours project. She was clearly well-versed in it and made the case that it would be worth writing the paper. My role was largely limited to writing the section on ‘prosody’ in the Introduction and the more discursive parts of the Discussion. She wrote the bulk of the paper, managing this during the first year of one of the autism projects while carrying out the duties outlined above. She selected the content, being entirely responsible for conducting the literature searches. She organised the layout, making useful innovations such as the tables detailing the important points of the papers. For each

paper, she made clear concise summaries of the main aims, methods and findings. That she had correctly gauged the need of the readership and produced a satisfactory piece of work is reflected in the fact that after submission only minor revisions were required and the piece was published in record time [2 months from submission to acceptance]. The paper is frequently cited in subsequent work on prosody in autism.

2. **McCann, J.,** Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Prosody and its relationship to language in school-aged children with high-functioning autism *International Journal of Language and Communication Disorders*, 42(6), 682-702

This paper was again largely of Joanne's own conceiving, and the writing and organisation of it almost entirely hers, considerably more so than in the previous publication. My role was largely limited to advising on the paragraph on 'prosody' in the section 'Prosody and Theory of Mind'. The paper was largely written while Joanne was carrying out all the interviewing for the prosody in autism projects, with attendant record-keeping, liaising with schools, teachers and parents. It reflects that Joanne had mastered the necessary techniques of statistical analysis and an ability to keep several strands of information in mind for the discussion section: the distinction between comparing chronological age-matched and language-matched controls with the autism group; the key features of autism (such as language, pragmatic and mentalising impairment), the competing approaches to modelling autism (weak central coherence, executive dysfunction etc.), and the relationship between prosody and language skills. Her writing style in this article shows development from mainly factual presentation to the making of closely argued points.

3. Peppé, S., **McCann, J.,** Gibbon, F. & O'Hare, A. (2007). Receptive and expressive prosodic ability in children with high-functioning autism. *Journal of Speech, Language, and Hearing Research*, 50(4), 1015-1028

This paper was largely written by me, with Joanne contributing or editing sections that touched on the relationship between prosody and language (and other authors reading and commenting). Joanne was also responsible for collecting the data and compiling it into usable spreadsheets. The whole paper benefited greatly from the inferences that could be made from her experience in conducting all the interviews with the 109 participants. In conducting these, Joanne showed a remarkable ability to encourage testees and keep them focused on the tasks while accommodating their difficulties.

4. **McCann, J.,** Peppé, S., Gibbon, F., O'Hare, A. and Rutherford, M. (2008). The Prosody-Language Relationship in Children with High-Functioning Autism. *In McGregor, E., Nunez, M., Cebula, K. and Gomez, J.C. (Eds.) Autism. An Integrated View from Neurocognitive, Clinical, and Intervention Research.*, Oxford, United Kingdom, Blackwell, 214-235

This chapter forms part of a book which was commissioned by Blackwell after a series of seminars held in Scotland by SARG (Scottish Autism Research Group). Presenters at the seminars included internationally recognised experts on autism. Only a handful were invited to write a chapter for the book, of which Joanne was one. My role was more limited than in the previous paper, consisting of a critical review of Joanne's report and suggesting amendments. This paper reflects essentially similar findings as in the last paper, with the addition of illustrative case studies. Joanne was responsible for selecting the content, particularly the in-depth analysis of the language and prosody of the two children featured in the case studies. The 'integrated view' of the title of the book reflects the feature that each chapter relates to others that treat related topics, and this constitutes a considerable section at the end of the chapter. In order to write this, Joanne had to have grasped the import of most of the other contributions to the book.

Down's Syndrome Papers

Statement from Sara Wood on behalf of all co-authors.

Joanne is currently a Research Fellow on a 3 year MRC funded (£348,000) longitudinal study entitled "Assessment And Treatment Of Impaired Speech Motor Control In Children With Down's Syndrome". She is part of a research team under the overall direction of Professor William Hardcastle (PI), Professor Jennifer Wishart and Dr Sara Wood.

Joanne's role includes the following:

- researching background literature for the project
- assessing 30 children with DS and 30 typically cognitively matched children on a range of speech, language and oromotor tasks
- designing and implementing therapeutic intervention for the DS group using both novel (EPG) and conventional techniques

- writing reports to parents and professionals summarising assessment findings and progress in therapy
- organising and maintaining contacts with participants
- keeping data records
- writing up papers reflecting findings and presenting project data at both international and national level in collaboration with other members of the research team.

5. **Cleland, J.,** Wood, S.E., Hardcastle, W.J., Wishart, J. & Timmins, C. (2010). The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. *International Journal of Language and Communication Disorders*. 45 (1), 83-95.

There are several studies which have looked at the type of speech errors produced by children with DS. Most studies have concluded that there is a delay in speech production skills. To date no studies explore the relationship between cognitive or language skills and intelligibility. In this paper the authors sought to determine whether the severity of the speech disorder correlates with language and cognitive level and to classify the types of errors that occurred in the speech of 15 children with DS.

This paper was predominantly written by Joanne. She was responsible for reviewing the appropriate literature, organising the layout, selecting an appropriate methodology for analysis (which was based on her earlier work in the field of autism) and for discussing the findings in light of current knowledge. Co-authors were responsible for reading the initial draft, commenting and suggesting changes to this and reading subsequent drafts where Joanne had reflected upon and taken on board comments from her colleagues. Following submission to the International Journal of Disorders of Communication the paper required minor revisions. The editors reported that "The reviews are in general favourable and suggest that, subject to amendments noted, your paper could be suitable for publication". The paper was resubmitted and has now been published. This was the first major joint article from the research team describing some of the results of the MRC project.

6. **Cleland, J.,** Timmins, C., Wood, S.E., Hardcastle, W.J. & Wishart, J. (2009). Electropalatographic therapy for children and young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23 (10), 787-801.

There are only 2 previously published studies looking at the effect of EPG therapy in children with DS (Gibbon, McNeil, Wood and Watson, 2003; Wood, Wishart, Hardcastle, Cleland and Timmins, 2009). The former is a single case study and the latter reports on two single cases. In this most recent publication the authors report in detail on 6 individuals with DS through both qualitative and quantitative analysis. The study provides support to the initial claims that EPG may be an effective means of intervention for improving speech intelligibility.

This paper was initially presented at an international conference, ICPLA2008 before being written for publication. As with the first paper, it was predominantly written by Joanne. Co-authors were responsible for reading the initial draft, commenting and suggesting changes to this and reading subsequent drafts where Joanne had reflected upon and taken on board comments from her colleagues, particularly with regard to statistics. Following submission to Clinical Linguistics and Phonetics the paper required some revisions which were discussed as a team. The paper was resubmitted and has now been published.

Sara Wood

December 2009

Appendix Four: Curriculum Vitae

Joanne Cleland

Academic Qualifications: BSc(Hons) Speech Pathology and Therapy, Queen Margaret University, 2002.

Membership of Professional Bodies: Member of the Royal College of Speech and Language Therapists and Health Professions Council registered.

Employment History

Company Name	Job Title	Start Date	End Date
Queen Margaret University	Research Fellow (FT)	Oct 2005	Current
Queen Margaret University	Research Assistant (PT)	May 2004	Oct 2005
NHS Lothian	Speech & Language Therapist (PT)	Aug 2004	Aug 2005
Queen Margaret University	Research Assistant (FT)	May 2002	May 2004

Publications

(Publications prior to 2009 are in my previous surname of McCann)

Peppé, S.J.E., Cleland, J., Gibbon, F.E., O'Hare, A.E., Martínez-Castilla, P. (2011). Expressive prosody in children with autism spectrum conditions. *Journal of Neurolinguistics*, 24, 41-53.

Cleland, J., Gibbon, F., Peppe, S., O'Hare, A. & Rutherford, M. (2010). Phonetic and phonological errors in children with high functioning autism and Asperger syndrome. *International Journal of Speech-Language Pathology*, 12, 69-76.

Cleland, J., Wood, S., Hardcastle, W., Wishart, J. & Timmins, C. (2010). The relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. *International Journal of Language and Communication Disorders*, 45, 83-95.

Cleland, J., Timmins, C., Wood, S., Hardcastle, W. & Wishart, J. (2009). Electropalatographic therapy for children and young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23, 926-939.

Timmins, C., Cleland, J., Wood, S., Hardcastle, W.J & Wishart, J. (2009). A perceptual and electropalatographic study of /ʃ/ in young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23, 911-925.

- Timmins, C., Cleland, J., Rodger, R., Wishart, J., Wood, S. & Hardcastle, W.J. (2009). Speech Production in Down Syndrome. *Down Syndrome Quarterly*, 11, 16-22.
- Wood, S., Wishart, J., Hardcastle, W.J., Cleland, J. & Timmins, C. (2009). The use of electropalatography (EPG) in the assessment and treatment of motor speech disorders in children with Down's syndrome: Evidence from two case studies. *Developmental Neuropsychology*, 12, 66-75.
- McCann, J., Peppe, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2008). The Prosody-Language Relationship in Children with High-Functioning Autism. In McGregor, E., Nunez, M., Cebula, K. and Gomez, J.C. (Eds.) *Autism. An Integrated View from Neurocognitive, Clinical, and Intervention Research*, (pp 214-235). Oxford, United Kingdom, Blackwell.
- McCann, J., Peppe, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Prosody and its relationship to language in school-aged children with high-functioning autism *International Journal of Language and Communication Disorders*, 42, 682-702.
- Peppé, S., McCann, J., Gibbon, F. & O'Hare, A. (2007). Receptive and expressive prosodic ability in children with high-functioning autism. *Journal of Speech, Language, and Hearing Research*, 50, 1015-1028.
- Peppé, S., McCann, J., Gibbon, F., O'Hare, A. & Rutherford, M. (2006). Assessing prosodic and pragmatic ability in children with high-functioning autism *Journal of Pragmatics*, 38, 1776-1791.
- McCann, J. & Peppé, S. (2003). Prosody in Autism Spectrum Disorders: A Critical Review. *International Journal of Language and Communication Disorders*, 38, 325-350.
- Peppé, S. & McCann, J. (2003). Assessing intonation and prosody in children with atypical language development: the PEPS-C test and the revised version. *Clinical Linguistics and Phonetics*, 17, 345-354.

Published Presentations/ Conference Proceedings

- McCann, J. & Wrench, A. (2007). A new EPG protocol for assessing DDK accuracy scores in children: A Down's Syndrome study. In J. Trouvain & W.J. Barry (Eds.), *Proceedings of the 16th International Congress of the ICPhS*, Germany (pp. 1985-1988). Dudweiler: Pirrot.
- Timmins, C., Hardcastle, W.J., Wood, S., McCann, J. & Wishart, J. (2007). Variability in fricative production of young people with Down's syndrome: an EPG analysis. In J. Trouvain & W.J. Barry (Eds.), *Proceedings of the 16th International Congress of the ICPhS*, Germany (pp. 1981-1984). Dudweiler: Pirrot.
- Hardcastle, W.J., Wood, S., Wishart, J., McCann, J. & Timmins, C. (2006). Electropalatography Therapy for Articulation Disorders in Children with Down Syndrome. *Down Syndrome Quarterly*, Canada, 8, 105.
- Gibbon, F., McCann, J., Peppe, S., O'Hare, A. & Rutherford, M. (2004). Articulation disorders in children with high functioning autism. *The International Association of*

Logopedics and Phoniatrics, Congress Proceedings, 29th August to 2nd September 2004, Brisbane, Australia.

Appendix Five: Self-Evaluation of Contribution to the Published Work

During my time at Queen Margaret University as a researcher I have worked on four research projects: two on prosody in Autism Spectrum Disorders and two on Electropalatography therapy for children with Down's syndrome. My interest in research initially stemmed from my undergraduate research project (McCann, 2002, "The ability of children with autism to use prosodic cues for phrasal interpretation"). As an undergraduate student I had observed several children with autism who presented with unusual intonation. This seemed to me to characterise these children, yet it was not part of the diagnostic criteria of the condition. I wondered if these children had a similar difficulty understanding intonation and designed an experiment inspired by Beach, Katz and Skowronski (1996) to investigate the understanding of prosodic cues at phrase boundaries. The methodology I developed was later used to make changes to the "Chunking" subtest of the PEPS-C assessment (Peppé & McCann, 2003). Upon completing my undergraduate degree, and thereby qualifying as Speech and Language Therapist I was appointed as a research assistant investigating prosodic skills in children with High-Functioning Autism. As I collected data from the children with HFA I formulated the hypothesis that those who did best at the prosody assessment had the best language skills. This observation sparked my interest in the interplay between speech/prosody and language and cognitive skills, leading to my subsequent collaboration with the senior colleagues whose knowledge, experience and plans were more compatible with these interests, thus providing the theme for my subsequent research. Thus my critical appraisal can draw on a broad but coherent track-record. What follows is a description of my journey as a researcher to date via the publications I have selected for this doctoral portfolio, with indicative citation counts from Google Scholar (24/08/2010).

1. **McCann, J.** & Peppé, S. (2003) Prosody in Autism Spectrum Disorders: A Critical Review. *International Journal of Language and Communication Disorders*, 38, 325-350.

My contribution: This paper is a critical review of the literature. I was responsible for searching for appropriate literature and critically appraising it, drawing together recommendations for future research. The recommendations made are addressed in papers 2 to 4.

Citations: 63

This paper was written early in my research career and was my first published paper. Its starting point was the literature I reviewed for my undergraduate research project, but in order to meet the requirements for publication I had to expand on this significantly in quantity and quality. Since this is a critical review, it is similar to the literature review in a traditional PhD, showing a critical and detailed knowledge of a substantial body of research. I was responsible for searching for appropriate literature and critically appraising it, drawing together recommendations for future research and bearing these recommendations in mind for our own research.

Writing this paper gave me the opportunity to become fully confident using databases such as Medline, a skill which has been invaluable throughout my research career. Writing for publication early on in my career gave me the opportunity to improve my writing skills and gain confidence in communicating with my academic peers. Since this was a critical review, rather than a research report, I became fully versed in the literature in my specialist area, providing a critical overview of the subject for both myself and my peers.

2. **McCann, J.,** Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2007). Prosody and its relationship to language in school-aged children with high-functioning autism. *International Journal of Language and Communication Disorders*, 42, 682-702

My contribution: This paper is based on data using the PEPS-C test. All data was collected, scored and analysed by myself and I was responsible for the writing and the generation of ideas in the discussion.

Citations: 17

This paper was my first research report. The idea for the paper came from my hands-on experience of collecting data from a large group of children with High-Functioning Autism. As I collected the data I observed that the children with better language skills tended to do better in the prosody assessment. I had also noticed that the literature profiling the speech, language and cognitive skills of a tightly defined group of children with HFA was sparse and therefore this paper was the ideal opportunity to add to the research evidence in this area.

Collecting data for this paper involved using a range of both standard (for example, standardised language assessments) and specialised research instruments (PEPS-C) with children with complex communication and behavioural needs. An earlier version of PEPS-C

had already been developed by Wells, Peppé and Goulondris (2004) but as part of the research project we substantially revised the test and computerised it. Details of changes to the test are outlined in Peppé and McCann (2003) and the changes were made on the basis of collaboration between Peppé and myself. Changes to the Chunking task were based on results from my own undergraduate project, showing my ability to both use and enhance and modify complex techniques and instruments. For the computerisation of the test I liaised with the software company, wrote scripts and piloted the new version of the test.

In order to confirm my hypothesis that prosodic ability was related to language ability I had to select and apply statistical tests and interpret the results. The results of this paper confirmed a relationship between prosody and language and I further hypothesised that this may be due to a difficulty with theory of mind in people with autism. This interpretation was my own and led to the research team applying for (and receiving) a grant to look at prosodic skills in children with Asperger syndrome. At this time, I also suggested that the new grant include measures of theory of mind alongside measures of language in order to try and disentangle the complex, and perhaps circular, relationship between language, prosody and theory of mind.

3. Peppé, S., **McCann, J.**, Gibbon, F. & O'Hare, A. (2007). Receptive and expressive prosodic ability in children with high-functioning autism. *Journal of Speech, Language, and Hearing Research*, 50, 1015-1028

My contribution: This paper is based on data I collected using the PEPS-C test. All data was collected and scored by myself. For this paper Peppé performed the analyses and did the majority of the writing. I contributed ideas to the discussion.
Citations: 34

As second author on this paper my role was mainly in the collecting and scoring of data and in commenting on drafts of the paper as part of the team. Working as part of a large research team has been both interesting and challenging. Over my time I have had the opportunity to work with and learn from a wide variety of researchers and clinicians senior to me. Moreover, as the clinical researcher responsible for collecting data from children with complex speech, language, cognitive and behavioural issues I have been uniquely placed to comment on the practical and ethical issues surrounding the research process.

4. **McCann, J.,** Peppé, S., Gibbon, F., O'Hare, A. & Rutherford, M. (2008). The Prosody-Language Relationship in Children with High-Functioning Autism. In *McGregor, E., Nunez, M., Cebula, K. and Gomez, J.C. (Eds.) Autism. An Integrated View from Neurocognitive, Clinical, and Intervention Research.*, Oxford, United Kingdom, Blackwell, 214-235

My contribution: This book chapter is based on data I collected using the PEPS-C test. All data was collected and analysed by myself and I was responsible for the writing and generation of ideas in the discussion. I selected the two children for case studies based on my in-depth knowledge of the study participants.

Citations: 1

During my research career I have been lucky enough to be part of special interest groups and national and international conferences. These have always provided important networking opportunities and the chance to hear the most up to date research. In 2003 researchers from across Scotland received funding from the British Psychological Society for a seminar series on current research in autism. Researchers were invited from both the UK and abroad to talk at seminars. I was one of the few researchers whose talk focused on language/communication research and was subsequently invited to submit a chapter for the above book. In part the chapter summarises the content of papers 2 and 3 but I felt that a book chapter was an ideal opportunity to reflect on my experience of meeting the individual children affected by autism by including two case studies of children whose language and prosody skills had interested me. Since finding a correlation between standardised measures of language and our prosody test I was interested in whether any of the children presented with impaired prosody in the face of normal formal language skills. I selected one child, "Iain", with normal language and one child, "Fiona" with impaired language.

As the member of the research team with the hands-on experience with the children I was uniquely placed to write case studies. Reporting such case studies emphasises the heterogeneity in autism, a finding which can often be neglected when reporting large group studies. Moreover, it gave me the opportunity to talk in detail about the prosody and language skills of these children, again something which is often not possible when reporting group studies.

The book that this chapter was published in was innovative in that the editors required that each contributor link his or her chapter to the others by writing a special

“integration” section. Since most of the other contributors were outside my field of research this was a challenging task, requiring me to synthesise new and complex ideas from areas outwith my previous knowledge.

Down’s Syndrome Papers

After two years as a full time researcher on the prosody in autism project I decided that I would like to increase my clinical skills and got a part-time post as a Speech and Language Therapist working with EPG and children with intractable speech disorders. Due to my work with prosody and my findings of delayed and disordered segments in autism, I became interested in speech disorders more generally. I took the opportunity to work on a clinical trial of EPG therapy for children with Down’s syndrome in order to both broaden my clinical experience and learn new research and technical skills. There are ways in which EPG is a change in direction, but as I learnt more about speech disorders in this population, I applied the knowledge from my previous work and it became clear to me that in general the reasons for delayed and disordered speech are not well understood, particularly regarding the interplay between cognitive, linguistic and anatomical/physiological constraints.

5. **Cleland, J.,** Wood, S.E., Hardcastle, W.J., Wishart, J. & Timmins, C. (2010). The relationship between speech, oromotor, language and cognitive abilities in children with Down’s syndrome. *International Journal of Language and Communication Disorders*. 45, 83-95.

My contribution: I contributed to refining the methodology for this paper by suggesting appropriate standardised assessments. All data was collected and analysed by myself and I was responsible for the writing and the generation of ideas in the discussion.

Citations: 4

While working with the families of children with Down’s syndrome, parents would often tell me how their child had poor speech but good cognitive ability or vice versa. Parents were often concerned that their severely unintelligible child was judged as severely cognitively impaired by strangers or that therapy focusing on speech was not provided because of the assumption that speech skills were commensurate with general cognitive skills. This indeed seemed to be the assumption in the literature, though there was no study to suggest that this had been proven empirically.

Using the methodology I had learnt in paper 2, I decided to write a paper investigating the relationship between speech, oromotor, language and cognitive skills in the children with Down's syndrome. A clear picture of no correlation between speech and language or cognition emerged. While it was then clear that delayed cognitive skills do not directly cause delayed/disordered speech production it remained unclear what the nature of the disorder was. From my clinical observation of children with Down's syndrome I felt that most children presented with impairments in both the phonological and motor speech systems, but that the primary diagnosis for most children with Down's syndrome is dyspraxia.

Early on in the Down's syndrome project I had noticed how difficult it was to collect good DDK data (diadochokinetic) from very young (aged around three) typical children and extrapolated that children with severe learning difficulties would experience the same issues. I therefore designed a DDK task specifically for young children or children with cognitive impairments (McCann & Wrench, 2007). The methodology of this task is my own creative response to this specific issue, but I sought help from Wrench in automating the labelling of the simultaneous EPG data.

6. **Cleland, J.,** Timmins, C., Wood, S.E., Hardcastle, W.J. & Wishart, J. (2009). Electropalatographic therapy for children and young people with Down's syndrome. *Clinical Linguistics and Phonetics*, 23, 787-801.

My contribution: I designed and implemented the therapy described in this paper and contributed to designing the outcome measures. All data was collected and analysed by myself and I was responsible for the writing and the generation of ideas in the discussion.

No citations to date

One of my major roles in the Down's syndrome project was designing and implementing individualised EPG therapy for children with Down's syndrome. This paper reports the progress in therapy for the first six children. As part of the therapeutic process I was responsible for selecting targets for intervention and I report on my method for doing so in this paper. Although the team had specified the word list for pre and post-therapy evaluations this proved inadequate for evaluating progress with production of velar stops. I therefore devised word lists to measure change in these consonants, recording them regularly during therapy sessions. I instigated the same process of devising target-specific word lists for each child based on their specific speech errors and these have proven to be invaluable.

Conclusion

The six selected publications and critical appraisal presented here represent my own personal journey from newly qualified Speech and Language Therapist/ Research Assistant to independent clinical researcher. Through publishing a critical review (paper 1), attending conferences and personal study I have gained a critical and detailed knowledge of speech and prosody in developmental disorders. Moreover, I have contributed to this research field by establishing a correlation between prosody and language, and by suggesting that this may be due to a deficit in theory of mind. In the field of learning disability, I have established that speech disorders in Down's syndrome are not related to cognitive ability and have shown that it is possible to remediate speech disorders in this population.

As a full-time externally funded researcher I have had the opportunity to gain hands on experience of data collection from a large number of both typically developing children and children with speech, language, cognitive and behavioural disorders. This experience has been instrumental in forming my research questions and driving my publications from both a theoretical and applied view point. These experiences have led me to develop new methodologies, such as my DDK assessment and to refine existing methodologies such as the PEPS-C assessment. I am now at a stage to submit a Research Council funding application with collaborators at other universities as a co-investigator and to forge my own clinical research agenda.